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Posterior Sagittal Anorectoplasty for a Recto-vaginal Fistula Occurring 30 Years after Swenson's Operation for Hirschsprung's Disease

Naoki OKUYAMA, Masayuki KUBOTA, Minoru YAGI, Satoru YAMAZAKI, Masahiro OHTANI and **Shinji TANAKA**

Department of Pediatric Surgery, Graduate School of Medical and Dental Sciences, Niigata University, Niigata, Japan

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Summary. **We herein report the case of a 37-year-old woman with a recto-vaginal fistula occurring 30 years after Swenson's operation for Hirschsprung's disease. As the patient had undergone multiple abdominal operations in childhood, a rectal pull-through operation was not indicated. The initial procedure of a direct closure of the fistula through the anal approach failed, and a sigmoid colostomy was performed at the same time. As a second procedure, a direct closure using a posterior sagittal approach was attempted and successfully performed. This approach provided a good operational field for performing a direct closure of the fistula. Two months after posterior sagittal anorectoplasty, a stoma closure was performed. The patient is now doing well without any recurrence of the fistula at 10 months after the operation. The experience of this case suggests that the posterior sagittal approach is a useful surgical option for rectal morbidities where abdominal and/or anal approaches are not easily performed.**

Key words — Recto-vaginal fistula, posterior sagittal approach, Hirschsprung's disease.

INTRODUCTION

A recto-vaginal fistula is a rare complication following previous surgical procedures for Hirschsprung's disease. The surgical treatment of this rare complication remains a matter of debate. It is difficult to close such fistulae using a vaginal or anal approach because the fistula can not be exposed with a good operational field. We herein report the case of a patient with a recto-vaginal fistula following Swenson's operation for Hirschsprung's disease which was successfully treated with a posterior sagittal anorectoplasty.

CASE REPORT

A 37-year-old woman presented with a 4-year history of a recto-vaginal fistula. At one month of age, the patient was diagnosed to have Hirschsprung's disease, for which a transverse colostomy was performed. At three years of age, she underwent Swenson's operation as a definitive procedure for Hirschsprung's disease. A colostomy closure was then performed at four years of age. Her post-operative course had been uneventful until a recto-vaginal fistula was found at 33 years of age. She initially underwent a direct closure of the fistula through a rectal approach with a sigmoid colostomy. However, the presence of a patent recto-vaginal fistula was confirmed

Correspondence: Naoki Okuyama, M.D., Department of Pediatric Surgery, Graduate School of Medical and Dental Sciences, Niigata University, 1-757 Asahimachi-dori, Niigata 951-8510, Japan.

Abbreviations–None.

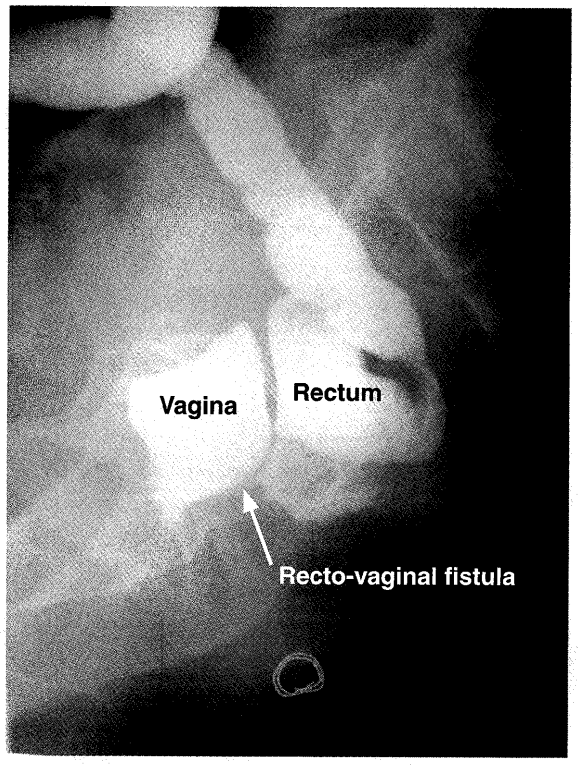

Fig. 1. A pre-operative barium enema shows a recto-vaginal fistula.

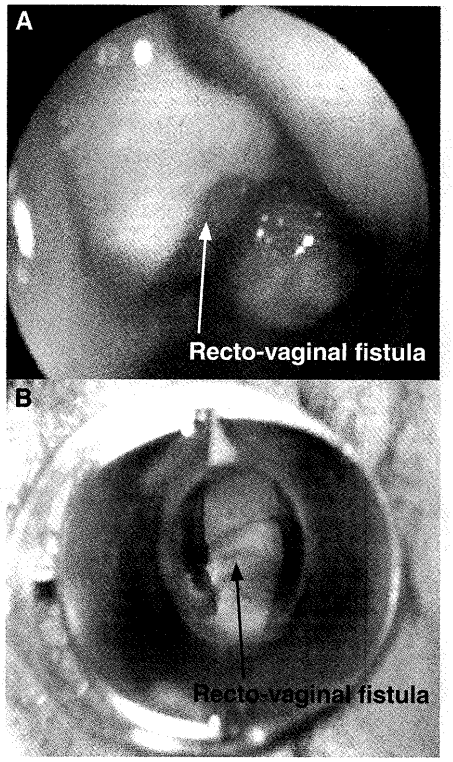

Fig. 2. A colono-fiber scope shows the recto-vaginal fistula **(A)** of the rectum wall and a vaginoscope shows the fistula **(B)** of the vaginal wall.

by a contrast study through the stoma at 36 years of age (Fig. 1). An endoscopic examination showed a fistula between the vagina and rectum, measuring about 3 mm in diameter and lying 6 cm above the dentate line of the anus (Fig. 2).

As this patient had undergone previous multiple abdominal surgical operations, a laparotomy or pull through method was abandoned because of possible dense adhesions in the abdominal and pelvic cavities. Therefore, the recto-vaginal fistula was directly closed using a posterior sagittal approach. The posterior rectal wall was opened and the fistula was thus exposed with a good operational field. The fistula was located in a thin fibrotic area without any mucosal lining. The fistula was therefore directly closed with purse string sutures, and thereafter the fibrotic region, including the fistula, was covered with an intact mucosa (Fig. 3). Ten days after the surgery, a barium enema was performed and no barium leakage was found (Fig. 4). Two months after posterior sagittal anorectoplasty, a stoma closure was performed. The patient is now doing well without any recurrence of the recto-vaginal fistula at 10 months after the operation.

DISCUSSION

Posterior sagittal anorectoplasty is a technique mainly used for the repair of high or intermediate type anorectal malformations. The main objective of this approach is to obtain a direct exposure of the anorectal region by means of a median sagittal incision that runs from the sacrum to the anal dimple while cutting through all muscle structures behind the rectum. Posterior sagittal anorectoplasty was first reported by Pena and De Vries[1,2] in 1962 and thereafter became accepted worldwide.

A posterior sagittal resection is also performed on patients with rectal aganglionosis as a primary surgery, as reported by Hedlund[3] in 1997. Lodz[4] reported that they performed a posterior sagittal abdominoperineal pull-through for patients with Hirschsprung's disease to decrease and eliminate the incidence of complications

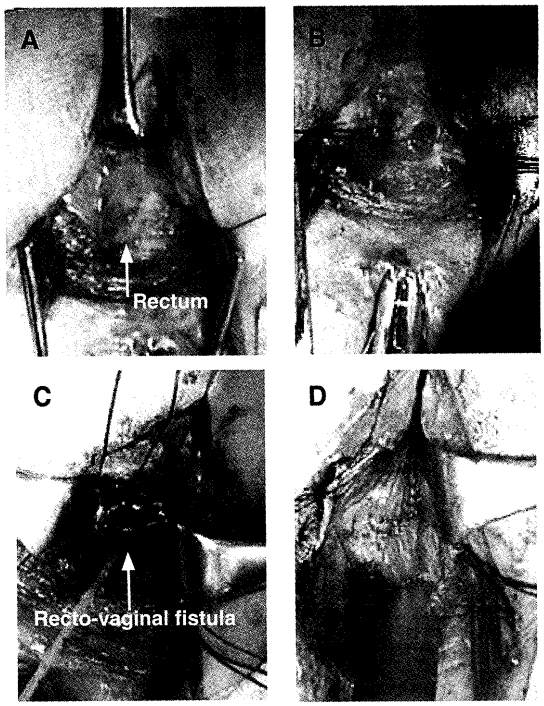

Fig. 3. The surgical strategy is shown in these pictures. **A.** A posterior sagittal incision was made on the skin and muscles. **B.** The posterior wall of the rectum was opened. **C.** The recto-vaginal fistula is shown with a good surgical view. **D.** The recto-vaginal fistula was thus closed directly.

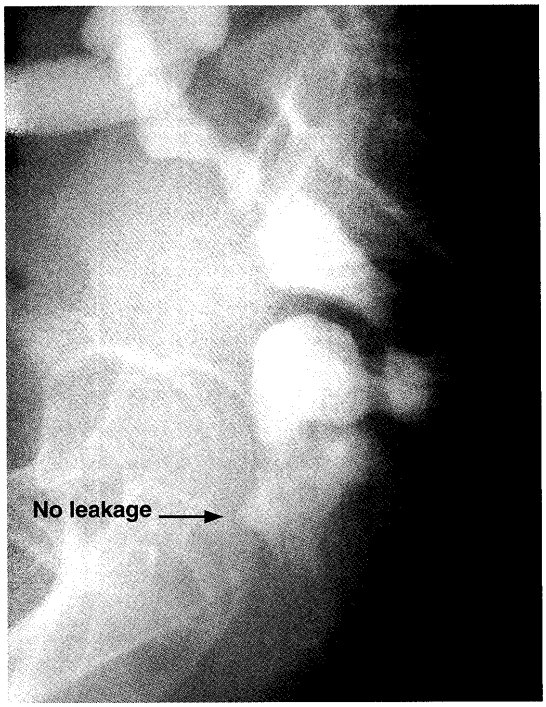

Fig. 4. A barium enema shows the fistula to be closed, and no incidence of barium leakage is observed.

related to major pull-through procedures, such as anastomotic stenosis, enterocolitis, constipation, and anastomotic leakage. Aggarwar et al.[5] reported that, in Hirschsprung's disease, a redo pull-through was indicated for anastomotic stricture, leakage, and retained aganglionosis after previous operations. The Duhamel or Swenson method has also been commonly used for redo operations. A pelvic dissection may be difficult, especially in Swenson's operation, because of fibrosis resulting from previous surgery or its complications. Aggarwal et al.[5] utilized a combined abdominal and posterior sagittal approach for a redo pull-through operation in Hirschsprung's disease and was successful. Kubota et al.[6] described the rectovaginal or rectourethral fistula as a troublesome complication after anorectal surgery because of dense adhesions around the fistula, and they therefore performed an endorectal pull-through using the posterior sagittal approach which proved to be successful.

Our patient had undergone multiple abdominal surgeries, and a direct closure for the recto-vaginal fistula with a rectal approach was performed for the patient three years earlier. Dense adhesion was presumed to exist around the fistula and the rectum, rendering it difficult to dissect the fistula or the rectum. Therefore, we opened the posterior wall of the rectum using the posterior sagittal approach, and this provided an excellent exposure of the operational field. Nevertheless, a pull-through of the rectum and colon was still difficult to perform, and the recto-vaginal fistula had to be closed directly.

The endorectal pull-through under a posterior sagittal approach, as described by Kubota et al., is considered to be a reliable method for a recto-vaginal fistula, but otherwise it is very difficult to perform because of severe adhesions– especially after previous surgery for Hirschsprung's disease. Compared with an endorectal pull-through, a direct closure of recto-vaginal fistula using a posterior sagittal approach is an easier and more useful method to perform when treating such patients. This is the first report of an application of the posterior sagittal approach to perform a direct closure of a recto-

vaginal fistula. Posterior sagittal anorectoplasty might therefore be a useful modality for the treatment of patients with rectal morbidity secondary to abdominal or pull-through operations.

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